Surgical Neurology International

SNI: Spine, a supplement to Surgical Neurology International

OPEN ACCESS

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A case of dumbbell-shaped epidural cavernous angioma in the lumbar spine

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Received: 23 April 15 Accepted: 08 May 15 Published: 25 June 15

This article may be cited as:

Yunoki M, Suzuki K, Uneda A, Yoshino K. A case of dumbbell-shaped epidural cavernous angioma in the lumbar spine. Surg Neurol Int 2015;6:S309-12. Available FREE in open access from: http://www.surgicalneurologyint.com/text.asp?2015/6/11/309/159378

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Abstract

Background: Most spinal cavernous haemangiomas occur in the vertebral body and purely extradural cavernous hemangiomas without any vertebral body involvement is rare and account for only 4% of all extradural spinal tumors. Dumbbell-shaped spinal cavernous angioma is extremely rare, only 10 cases have been reported in the literature.

Case Description: A 77-year-old female presented with a one-year history of lumbago and right-sided L3 dermatomal hypoesthesia. A dumbbell mass at the L2/3 vertebral level was identified on lumbar MRI. The lesion was irregularly shaped and isointense on T1W and hyperintense on T2W and DWI images with homogenous contrast enhancement. A presumptive diagnosis was schwannoma, but other malignant neoplasms were also considered because of its irregular shape, minimally dilated neural foramen and the involvement of the non-enhanced L3 nerve root. The patient underwent surgery with a lateral extracavitary approach. A histopathological examination revealed cavernous hemangioma.

Conclusion: Cavernous hemangioma should be included in the differential diagnosis of dumbbell-shaped spinal tumors when the intervertebral foramina is not highly dilated and non-enhanced nerve root is identified in the tumor.

Key Words: Cavernous hemangioma, differential diagnosis, dumbbell, epidural, lumbar



INTRODUCTION

Most spinal cavernous hemangiomas occur in the vertebral body and may sometimes extend into the epidural space. Purely extradural cavernous hemangiomas without any vertebral body involvement are extremely rare, and account for only 4% of all extradural spinal tumors. We herein present a rare case of a dumbbell-shaped epidural cavernous hemangioma at the level of L2/3. This case is reported because of its

rarity, the unusual dumbbell shape of the lesion and the difficulty in making a preoperative diagnosis.

CASE REPORT

A 77-year-old female presented with a 1-year history of lumbago and right-sided L3 dermatomal hypoesthesia. Her muscle strength and deep tendon reflexes were normal. Magnetic resonance imaging (MRI) of the lumbar spine revealed an irregularly-shaped, well-circumscribed

paraspinal mass with minor intraspinal extension through the L2/3 intervertebral foramen. The lesion was isointense on Tl-weighted (TlW) and hyperintense on T2W and diffusion-weighted images, with strong homogenous enhancement in a gadolinium (Gd) contrast study [Figure 1]. In the Gd-enhanced images, involvement of the nonenhanced right L3 nerve root in the tumor was identified [Figure 1d and e]. Computed tomography (CT) demonstrated mild enlargement of the right neural foramen at L2-3, but there were no remarkable erosive changes of the vertebral body, pedicle or lamina [Figure 2a and b]. All modalities of sensations (pin prick, touch, temperature, vibration) at the L3 dermatomes on the right side were observed. The bilateral lower limb power was normal. The results of the general examination were unremarkable, and no sensorimotor deficits were detected in the upper limbs. A presumptive diagnosis was schwannoma, but the possibility of a neurofibroma or other malignant neoplasm was considered because of the tumor's irregular shape, lack of an enlarged neural foramen and the involvement of the nonenhanced L3 nerve root. The patient underwent surgery with a lateral extracavitary approach.[18]

A longitudinal right paravertebral skin incision 15 cm in length at the L1–L3 level was made, followed by clear identification of the lumbodorsal fascia. The fascia was then incised in line, and a clear plane was identified between the multifidus medially and the longissimus laterally. The muscles were meticulously teased apart. The transverse processes of Th2 and Th3 were identified by fluoroscopy [Figure 3], and were exposed with monopolar cautery. A Th3/4 external foraminotomy was performed and the transverse processes of Th2 and Th3 were

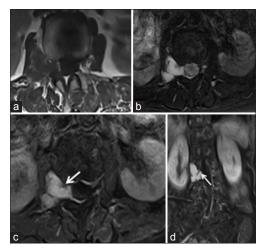


Figure 1: Preoperative magnetic resonance images at the level of L2/3 showing an irregularly-shaped, well-defined extradural lesion with paraspinal transforaminal extension that was hypointense on TI-weighted (TIW) (a) and hyperintense on T2W (b) and diffusion-weighted images. (c) Contrast-enhanced TIW images of transverse (d) and coronal sections showed the right L3 nerve root ventrally separated from the homogeneously enhanced tumor (arrow)

removed, then a brownish-red, highly vascular mass was identified [Figure 3]. The tumor was highly vascular and bled heavily upon the acquisition of a small biopsy. The capsule was coagulated to control the hemorrhage. An intraoperative histopathological examination indicated that the tumor was a hemangiomatous neoplasm, without evidence of malignancy. The tumor extended into the intrathecal region through the L2/3 neural foramen. The right L3 nerve root was identified in the ventral part of the tumor. The tumor adhesion to the nerve root was severe; therefore, we left the tumor around the nerve root behind due to concerns about possible nerve root damage. The tumor was removed subtotally. The patient's postoperative course was uneventful. She was discharged 2 weeks after surgery. The histological examination of the tumor showed it to be a cavernous hemangioma [Figure 3].

DISCUSSION

Hemangiomas are congenital vascular malformations whose pathologies are considered to be hamartomatous malformations. [3,7] They are classified based on the predominant type of vascular channel (capillary, cavernous, arteriovenous or venous) observed during the histological examination. Spinal epidural hemangiomas account for 4% of all spinal epidural tumors, mostly occurring as a primary lesion in the vertebral bone. [2] Some authors have reported cases of purely epidural hemangiomas, most of which were of the cavernous type.^[3,13] In contrast to the brain or spinal cord hemangiomas, the patterns of density on CT scans and signal intensity on MRI are more homogeneous in epidural hemangiomas. On CT studies, these neoplasms appear as intermediate or slightly hyperdense extradural masses. By MRI, they display high signal intensity on T2W images, which may be explained by the high content of stagnant blood. Slow blood flow may contribute substantially to the signal, conferring a low or intermediate signal intensity on TIW images,

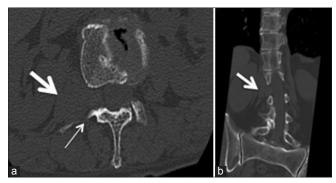


Figure 2: (a) A transverse computed tomography (CT) scan at the level of L2/3 showing a left-sided dumbbell-shaped mass (thick arrow). The posterior wall of the right intervertebral foramen was slightly eroded (thin arrow). (b) A coronal CT scan demonstrating the mildly enlarged right intervertebral foramen and dumbbell-shaped mass (thick arrow)

although there is no direct anatomical relationship with the intervertebral disk or exiting nerve root. [2,4]

About 80% of spinal epidural cavernous angiomas are in the thoracic spine, most of which are located posterior within the spinal canal and present with myelopathy. [2] In cases of lumbar epidural cavernous angiomas, they tend to be located in the ventral extradural space and cause lumbar radiculopathy, clinically and radiologically

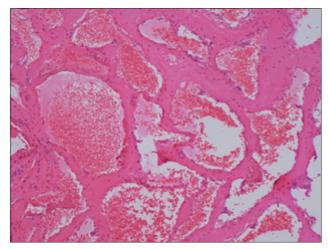


Figure 3: A microscopic examination revealed numerous dilated vascular channels of variable sizes lined by a single layer of flattened epithelial cells that was filled with blood elements. The tumor stroma consisted of typical fibrous tissue. The histological findings were characteristic of cavernous hemangioma. (H and E, ×100)

mimicking the presence of disk herniation. [15] These characteristics were also observed in 10 previously reported cases of dumbbell-shaped extradural cavernous angiomas [3,5,6,8,9,11,12,14,15,17] [Table 1]. Seven of the 10 cases were in the thoracic spine and three were in the lumbar spine. The symptoms of the dumbbell-shaped hemangiomas in the thoracic spine were mainly myelopathy, while those in the lumbar spine mainly led to radicular pain.

Dumbbell-shaped spinal tumors are usually thought to be schwannomas, neurofibromas or meningiomas. [10] Most of these tumors have enlarged intervertebral foramina, whereas cavernous angiomas do not tend to be dilated [10,12] [Table 1]. Some authors have reported that 10–40% of dumbbell-shaped spinal tumors are malignant tumors such as lymphoma, metastatic tumors, sarcoma, etc., which also tend to lack a dilated intervertebral foramina. [10,12] Cavernous hemangioma and malignant tumors should therefore be included in the differential diagnosis of dumbbell-shaped spinal tumors when the intervertebral foramina is not highly dilated. [10]

It has been suggested that a complete excision of the cavernous angioma is related to a favorable outcome. In the present case, however, we left a small amount of the tumor around the nerve root behind due to concerns about possible nerve root damage. When complete removal is not possible, radiosurgery is increasingly becoming an option, as advances in radiosurgical equipment are enabling safe and

Table 1: The reported cases of dumbbell-shaped epidural spinal hemangiomas

Author	Age/Sex	Level	Symptom	Surgery	MR findings			Descripion as to Intervetebral
					T1	T2	T1-Gd	foramen
Rovira 1999	51/W	L3/4	Sciatica	Laminectomy	ISO	High	HE	Not mentioned, but not dilated in CT image
Franz 1987	23/M	T3/4	Spastic paraplegia	Laminectomy Total removal	-	-	-	Dilated
Morioka 1986	50/M	T2/3	Hypesthesia below chest	Thoracotomy and laminectomy Total removal	-	-	-	Not dilated
Haimes 1991	46/M	T3/4	Hypesthesia of bilateral feet	Thoracotomy and laminectomy Partial resection	ISO	High	-	Mildly dilated
Uchida 2010	75/M	T11/12	Progressive paraparesis	Laminetomy Total removal	ISO	High	HE	Not mentioned, but not dilated in MR image
Lanotte 1994	27/F	T1/2	Paresthesi in bilateral leg	Arthrolaminectomy Total removal	Slightly low	High	HE	Dilated
Feider 1991	50/M	L3/4	Sciatica	Total removal	ISO	High	IE	Not mentioned, but not dilated in MR image
Padovani 1982	75/M	T3-6	Gait disturbance	Laminectomy Partial removal	-	-	-	Not dilated
Fukushma 1987	54/M	T7/8	Spastic paraparesis	Laminectomy Total remval	-	-	-	Not mentioned, but not dilated in CT image
Harrington 1995	37/F	L3/4	Leg numbness and pain	Hemilaminectomy Total removal	ISO	-	IE	Not dilated

HE: Homogenously-enhanced, IE: Irregularly-enhanced

accurate targeting of lesions.^[1,16] We are now considering adjuvant radiosurgery for the residual tumor.

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